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Quality of life of mothers of children with myelomeningocele

Jakość życia matek dzieci z przepukliną oponowo-rdzeniową

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ABSTRACT

Introduction: Children with myelomeningocele (MMC) require complex treatment and long-term commitment from the family in the therapeutic and educational process. All the burdens of the chronic disease affect the quality of life of parents and guardians. The aim of this study to assess the quality of life of parents of children with MMC. **Material and methods:** The study included 50 mothers of children with MMC. The WHOQOL-BREF questionnaire was used as a research tool; it enables obtaining a quality of life profile in four domains areas: physical health, psychological, social relationships, and environment. The child's motor impairment was assessed using the classification of Hoffer. **Results:** Significant differences were observed in all domains in the assessment of quality of life (physical health, psychological, social relationships, and environment). Mothers of boys with MMC from the villages reported a better quality life in the psychological domain Mothers of girls with MMC reported a better quality life in the physical health domain. Mothers of healthy children better assessed the individual overall perception of quality of life (82.4 ± 9.6) than parents of children with MMC (74.4 ± 11.5) and overall perceptions of their own health (parents of healthy children (78 ± 11.6); parents of children with MMC (74 ± 14.1). **Conclusions:** Mothers of children with MMC had a lower quality of life in all the analyzed domains compared with mothers of healthy children. Deterioration in the quality of life of mothers with sick children is more common among those living in the city.

Introduction

Myelomeningocele (MMC) is the most common neurological congenital anomaly, affecting approximately 300 000 newborns worldwide every year. The incidence is approximately 1 case per 1000 in the US and ranges from 0.7 in central France to 7.7 in the United Arab Emirates, and 11.7 in South America [1]. In the United Kingdom and Ireland, yearly prevalence of neural tube defects declined, predating any periconceptional folic acid supplementation policy initiatives, from 45 per 10 000 births in 1980 to 10 to 15 per 10 000 in the 1990s [2]. In contrast, in the rest of Europe the prevalence during the 1980s and thereafter was close to 10 per 10 000 births. In Europe (excluding Southern Europe), in spite of the significant decrease in neural tube defects

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prevalence in Northern Netherlands, the decrease for all registries combined is slight and non-significant was found. In South Europe the decline in neural tube defects prevalence since 1992 was significant [3].

The prevalence of MMC in Poland is 6.2 per 10 000 births [4]. Mejnartowicz determined the neural tube defects prevalence in children born in 1997–2002 to mothers residing in the Wielkopolskie, Kujawsko-Pomorskie and Lubuskie Provinces [5]. The calculated neural tube defects prevalence was 10.87 per 10 000 live- and stillbirths in all three provinces. In the same period, the prevalence among liveborns was 10.12, and among stillborns 125.76 per 10 000. The prevalence of different types of neural tube defects per 10 000 live- and stillbirths was as follows: anencephaly 2.35, spina bifida 7.27, encephalocoele 1.25.There were no significant differences between the results of the current study and the results of the previous polish population-based studies.

Patients with MMC have a range of physical (the difficulties with bowel and bladder management as well as ambulation challenges), intellectual, and communication impairments, with a wide range of severity. All clinical symptoms of MMC have a significant and cumulative impact on family functioning [6].

The chances of survival for children with a severe form of MMC have increased significantly, and medical care shifted the emphasis from life-saving interventions to promoting quality of life for these children and their families. According to Vermaes et al. [7], little is known about the impact of the disease on family functioning. MMC is the second most common birth defect in the world. Its occurrence depends on the geographical region, genetic and environmental factors [8]. The diagnosis introduces anxiety and a sense of unpredictability in the parents' lives. Often parents feel lonely in the fight against the disease; they lack systemic support. Achilles et al. [9] found that parents of children with disabilities face many challenges in psychological adaptation, much greater than parents of healthy children, in particular if the disabled child has more than one disability. The degree of disability in MMC depends on the location of spinal cord segment damage and type of defect (MMC *tectum, apertum*). Since the mid – 1960s, early surgical treatment of spina bifida increased the survival rate of children with severe cases of spina bifida, and in recent years the development of prenatal treatment at approximately 20 weeks of pregnancy has further improved the chances for survival [9]. As a result, medical workers were given the task of supporting the quality of life for these children and their families. On the one hand, improvement of the quality of life depends on medical actions (*e.g.*, urological, orthopedic, degree of hydrocephalus); on the other hand, on psychosocial actions, depending on the development of science associated with the chronic disease [10–12].

The concept of quality of life infiltrated from everyday language to science, which is why, despite the universality of its application, it is difficult to define. The WHO defines quality of life as individuals' perception of their life situation in the cultural context, value system in relation to the environmentally conditioned tasks, expectations and standards. It is a comprehensive evaluation method of an individual's physical health, emotional state, self-reliance, degree of independence from their surroundings, as well as the relationship with the environment and personal beliefs [13, 14]. In medicine, there is a concept of quality of life conditioned by health status (Health Related Quality of Life; HRQOL). It is a functional effect of disease and its treatment experienced by the patient [14]. Quality of life is important in medical practice in order to improve the doctor–patient relationship, to evaluate the effectiveness and relative merits of different treatments in the evaluation of health services, and in research and health policy development [13, 14].

The World Health Organization Quality of Life (WHOQOL-BREF) instrument comprises 26 items, which measure the following broad domains: physical health, psychological health, social relationships, and environment. The WHOQOL-BREF instrument was developed collaboratively in a number of centres worldwide, and has been widely field-tested [14–18].

To our knowledge, few studies have examined the WHOQOL-BREF of parents of children with MMC. The aim of this study was to assess the quality of life of parents of children with MMC.

Methods

Design

A cross-sectional questionnaire survey approaching children and adolescents registered with a MMC diagnosis at the Department of Pediatric Rehabilitation, Children's University Hospital in Białystok, Poland. The survey was conducted between November 2011 and July 2012. The study included 50 mothers of children with MMC, who were sent the WHOQOL-BREF questionnaire. The questionnaire was filled at home by 50 mothers of children with MMC.

Sample and recruitment

Out of the 91 eligible children identified through clinical appointment schedules, 50 (55%) parents agreed to participate in the study. Children with MMC comprised 27 (54%) girls and 23 (46%) boys. Mean age of the children was 10.02 ± 4.54. Fifty percent of respondents lived in the city, 50% in the country, 31 (62%) of mothers did not work, 31 (62%) patients had a secondary education, 11 (22%) higher, and 8 (16%) primary.

The control group consisted of 50 parents of healthy children. The study group consisted of 27 (54%) of girls and 23 (46%) of boys. Forty-seven percent of the respondents were from the city and the 53% from the country. Parents of healthy children had similar education. Mean age of the children was 8.70 ± 3.65 years.

Motor function

The ambulatory function in patients with MMC was defined according to Hoffer et al. [19] as 4 categories community, household, nonfunctional, and nonambulators scored 4-1. Hoffer's classification: 1 nonambulators; 2 nonfunctional ambulators; 3 household ambulators; 4 community ambulators. The MMC level was defined as the lowest level on the better side at which the child was able to perform an

antigravity movement through the available range of joint motion.

The WHOQOL-BREF questionnaire

The research tool was the WHOQOL-BREF questionnaire (World Health Organization Quality of Life BREF), Polish version. Assessment Instrument: short version, which contains 26 questions divided into four domains: **D1**. *Physical health*: general health assessment, pain and discomfort, dependence on medication and medical care, energy and fatigue, sleep and rest, ability to work and perform daily living tasks, and mobility. **D2.** *Psychological*: perception of own body, positive and negative feelings, self-esteem, personal beliefs, spirituality, religion, thinking, learning, memory and concentration. **D3**. *Social relationships*: personal relationships, received social support, and sex life. **D4**. *Environment*: freedom, security, surroundings, physical environment, communication, finance, information, access to health and social services, and spare time. Each question is assigned a corresponding number of points from 1 to 5, and the patient has to choose from the following possible answers: 1 point – very dissatisfied, 2 points – dissatisfied, 3 points – neither satisfied nor dissatisfied, 4 points – satisfied, 5 points – very satisfied. The scale includes items (questions) which are analyzed separately: **Question 1**: concerning the individual overall perception of quality of life; **Question 2**: concerning the individual general perception of health [14, 16].

Analyses of internal consistency, discriminant validity, and construct validity suggest the WHOQOL-BREF is a psychometrically strong measure of quality of life. Cronbach's alpha ranged from 0.63 (social relationships) to 0.76 (physical health) in this research. The measure has been used internationally to research subjective quality of life in individuals with myelomeningocele.

The study was approved by the Bioethics Committee of the Medical University of Białystok. All subjects gave informed consent to complete the questionnaire.

Statistical analysis

The data were analyzed with the statistical package Statistica v. 7.1. Descriptive statistics including mean and standard deviations were used for sample characteristics. When comparing 2 groups, the chi-square test for nonordered categorical variables was used. The *t*-test was used for comparison values of the quality of life between groups. Spearman's analysis was used to measure the dependence of mothers of quality of life and the motor function of patients, working status and education level. A value of $p < 0.05$ was considered statistically significant.

Results

The studied groups were comparable (no significant difference) in terms of age, sex, education and residence.

Due to the locomotor level according to Hoffer, 31 (62% of children with MMC were nonambulators (require a wheelchair), 5 (10%) of the children were nonfunctional ambulators (require assistance to walk), 3 (6%) of the children were household ambulators (able to walk at home), and 11 (22%) of the children were community ambulators (no limitations).

Table I – Concomitant symptoms of the disease in children with MMC $n = 50$ reported by the mother
Tabela I – Objawy współistniejące z chorobą u dzieci z MMC $n = 50$ relacjonowane przez matkę

Symptoms	Problem occurred
Neurogenic bladder (catheterization)	96%
Orthopedic problems	63.83%
Difficulty concentrating	34.0%
Learning difficulties	27.66%
Speech problems	19.15%
Vision disorders	14.89%
Allergy	12.77%
Behavioral disorders	6.38%
Mental retardation	6.38%
Impaired hearing	6.38%
Sleep disorders	4.25%
Panic attacks	4.30%
Rheumatoid diseases	2.13%
Epilepsy	2.13%
Chronic respiratory diseases	2.13%

MMC – myelomenigocele.

An interview with mothers of children with MMC found that most problems with the child concerned neurogenic bladder (96%), orthopedic problems (64%), problems with concentration (34%), and with learning (28%). Details are shown in Table I.

Comparing the responses of mothers of children with MMC with the control group of mothers of healthy children, we observed statistically significant differences in all four domains (physical health, psychological, social relationships, and environment). Comparing the data in Table II, the greatest differences were in the physical health domain $p = 0.004$ and psychological domain $p = 0.008$.

In the assessment of the quality of life by mothers of children with MMC, we found no statistically significant differences based on sex (boys, girls). Details are presented in Table III.

Due to the place of residence of mothers of children with MMC the largest difference was observed in the physical health domain – a statistically significant result (mothers from the country D1 – 23, mothers from the city D1 – 21.4);

Table II – Comparison of quality of life of mothers of children with MMC on the WHOQOL-BREF scale with a group of healthy children $n = 50$
Tabela II – Porównanie jakości życia matek dzieci z MMC w skali WHOQOL-BREF do grupy dzieci zdrowych $n = 50$

Domains	MMC $n = 50$	Controls $n = 50$	*P* value
Physical health	22.2 ± 2.23	23.44 ± 1.95	0.004
Psychological	21.42 ± 2.08	22.64 ± 2.45	0.008
Social relationships	11.32 ± 1.71	12.04 ± 1.59	0.031
Environment	27.68 ± 4.22	29.24 ± 3.84	0.056

MMC – myelomenigocele.

Table III – Comparison of quality of life of mothers of children with MMC based on sex on the WHOQOL-BREF scale
Tabela III – Porównanie jakości życia matek dzieci z MMC w zależności od płci w skali WHOQOL-BREF

Domains	Mothers of children with MMC		
	Boys $n = 23$	Girls $n = 27$	P value
Physical health	22.13 ± 1.6	22.26 ± 2.68	0.841
Psychological	21.44 ± 1.7	21.41 ± 2.39	0.963
Social relationships	11.17 ± 1.56	11.44 ± 1.85	0.581
Environment	27.78 ± 3.75	27.59 ± 4.65	0.875

MMC – myelomenigocele.

Table IV – Comparison of quality of life of mothers of children with MMC based on place of residence (city, country) on the WHOQOL-BREF scale
Tabela IV – Porównanie jakości życia matek dzieci z MMC w zależności od miejsca zamieszkania (miasto, wieś) w skali WHOQOL-BREF

Domains	Mothers of children with MMC		P value
	Country $n = 25$	City $n = 25$	
Physical health	23.04 ± 1.99	21.36 ± 2.18	0.003
Psychological	21.72 ± 2.03	21.12 ± 2.13	0.312
Social relationships	11.44 ± 1.73	11.2 ± 1.71	0.624
Environment	27.04 ± 4.42	28.32 ± 4.0	0.288

MMC – myelomenigocele.

while in other domains no statistically significant differences were observed (Table IV).

In the assessment of the quality of life of mothers of children with MMC living in the city or in the country, we found no statistically significant differences based on the sex of children (data are not shown).

Mothers of boys with MMC living in the country reported significantly higher quality of life scores compared with mothers living in the city in the psychological domain (Table V).

In the assessment of the quality of life of mothers of girls with MMC based on place of residence, we obtained statistically significant results in the physical health domain.

Table V – Comparison of quality of life of mothers of boys with MMC living in the city and mothers of boys with MMC living in the country on the WHOQOL-BREF scale
Tabela V – Porównanie jakości życia matek chłopców z MMC mieszkających w mieście z matkami chłopców z MMC mieszkających na wsi w skali WHOQOL-BREF

Domains	Mothers of boys with MMC		P value
	Country $n = 12$	City $n = 11$	
Physical health	22.75 ± 1.22	21.46 ± 1.75	0.051
Psychological	22.25 ± 1.60	20.55 ± 1.37	0.012
Social relationships	11.58 ± 1.31	10.73 ± 1.74	0.194
Environment	26.42 ± 3.99	29.27 ± 2.97	0.066

MMC – myelomenigocele.

Table VI – Comparison of quality of life of mothers of girls with MMC living in the city and mothers of girls with MMC living in the country on the WHOQOL-BREF scale
Tabela VI – Porównanie jakości życia matek dziewczynek z MMC mieszkających w mieście z matkami dziewcząt z MMC mieszkających na wsi w skali WHOQOL-BREF

Domains	Mothers of girls with MMC		P value
	Country $n = 13$	City $n = 14$	
Physical health	23.31 ± 2.53	21.29 ± 2.53	0.048
Psychological	21.23 ± 2.32	21.57 ± 2.53	0.719
Social relationships	11.31 ± 2.10	11.57 ± 1.65	0.718
Environment	27.62 ± 4.87	27.57 ± 4.62	0.980

MMC-myelomenigocele.

Table VII – Correlations between quality of life of mothers on the WHOQOL-BREF scale scores and ambulatory function of patients, not working, and level of education
Tabela VII – Korelacje pomiędzy jakością życia matek dzieci z MMC w skali WHOQOL-BREF a funkcją motoryczną pacjentów, pracą i poziomem wykształcenia matek

Domains	Ambulatory function	Not working	Level of education
Physical health	−0.20658	0.16537	−0.11122
Psychological	−0.14039	0.31782[a]	−0.04721
Social relationships	−0.16877	0.16964	−0.09417
Environment	−0.08734	0.23254	−0.20552

[a] $p = 0.024498$, R = Spearman rank correlation coefficient.

Mothers of girls from the countryside D1 – 23.3, mothers of girls from the city D1 – 21.3. Details are shown in Table VI.

The questions analyzed separately show that mothers of healthy children significantly ($p = 0.02$) better assess individual overall perception of quality of life (82.4 ± 9.6) compared with mothers of children with MMC (74.4 ± 11.5). The question regarding individual general perception of own health was also better (not significant) assessed by mothers of healthy children (78 ± 11.6) compared with mothers of children with MMC (74 ± 14.1). No significant correlations between the ambulatory function of patients with MMC and quality of life of mothers on the WHOQOL-BREF scale were found (Table VII).

Discussion

Our study indicates that mothers of children with MMC have a reduced perception of quality of life in all the analyzed domains compared with mothers of healthy children. No gender differences were found in reports of the WHOQOL-BREF. Mothers of children with MMC living in the city reported lower quality of life compared with mothers living in the country.

These results are comparable with findings of other studies [7, 11, 20, 21]

Our findings are particularly important for clinicians involved in the management of children with MMC. When a child is born with a disability, in addition to rapid adaptation, the family has to cope with stress, sadness,

disappointment and challenges, which can lead to a serious crisis and even disrupt family life. Parents should analyze the child's development, use regular comprehensive rehabilitation, maintain contact with a number of specialists and numerous social institutions or services. They are often faced with important decisions to be made for the benefit of the disabled child and economic decisions that affect the family. A child born with a disability is always a tragedy for guardians, but early specialist intervention and adequate financial support helps in adjusting and in positive commitment in the care and development of the child, even if the child is different and requires special treatment [20–22]. In this study, we found that most children had orthopedic problems (64%), problems with concentration (34%) and learning (28%). Comparing the responses of mothers of children with MMC with the group of mothers of healthy children, we observed statistically significant differences in all four domains (physical health, psychological, social relationships, and environment). The largest differences occurred in the physical health and psychological domains. Furthermore, mothers of healthy children better assessed their individual general perception of quality of life and general health compared with mothers of children with myelomeningocele. There are many studies in the literature evaluating the quality of life of children with chronic diseases such as autism or mental retardation [23–26]. There are few studies evaluating the quality of life of mothers, in particular of children with MMC [7, 21].

Diego Mugno et al. [23] evaluated the quality of life among parents of patients with different types of disability: Pervasive Development Disorder, cerebral palsy and mental retardation compared with a control group, and compared the quality of life for mothers and fathers. Compared with parents of healthy children, parents in the Pervasive Development Disorders group reported significantly decreased physical activity, and social relations, and individual overall perception of quality of life, and health. Parents of children with Pervasive Development Disorders showed higher loads for a combination of environmental and genetic factors. Schieve [24] also stresses that parents of children with developmental disabilities may experience severe stress, impaired physical functioning, fatigue or exhaustion. We found that based on the place of residence of mothers of girls with MMC the largest differences were in the physical health domain. Mother of girls from the country better evaluated the physical health domain compared with mothers of girls from the city. However, based on the place of residence of mothers of boys with MMC, the largest differences were observed in the psychological domain. Similarly, Weiss [26] stressed that more attention should be paid to the needs of parents (especially mothers). Social support and different coping strategies in the face of illness of a child with a disability should be tailored to respond positively to changing individual needs. Vitaliano et al. [27] emphasized that the level of loss of quality of life in families of children with severe chronic disease may be determined by environmental factors such as socio-economic status and social support. In this study, statistically significant differences occurred in the environmental domain compared with the control group of healthy children. Vermaes et al. [7] reported that a MMC diagnosis initially provokes traumatic stress symptoms in three-quarters of the parents, but in most of them, these symptoms decrease during the first 4 years of the child's life. Among a small group of parents these severe symptoms of stress persist beyond school age. Professional psychological help may be needed for this group of parents whose stress levels do not decrease after preschool. The severity of physical dysfunction of a child with MMC enhances stress among parents, especially problems associated with neurogenic bladder and constipation in school age. The studied group of mothers reported mainly bladder and orthopedic problems, difficulty concentrating, and problems with learning. In another report Vermaes [18] et al. showed major negative effects of MMC on the parent–child relationship (parent stress and over-protectiveness) and on the psychological situation of the caregivers, especially mothers. In the assessment of the quality of life of mothers of boys with MMC, based on place of residence, we obtained statistically significant results in the psychological domain. In another study, [13] stress management, parenting skills, relationship with the partner, family atmosphere and environmental factors were found to be associated with changes in the psychological self-regulation of parents. Furthermore, mothers with more supportive families and marriages and less conflicted reported lower levels of psychological symptoms. The study by van't Veer et al. [28] indicates that analysis of quality of life of parents of children with MMC sets the direction for state economic and educational activities for people with disabilities.

Study limitations

The size of the study group – 91 mothers, of which only 50 (55%) completed the survey. Only mothers were studies because they were mainly involved in the therapy of their children. We did not study socioeconomic factors. We are going to expand our sample on the patients and their fathers.

Conclusions

Mothers of children with MMC had a lower quality of life in all the analyzed domains compared with mothers of healthy children. Analysis of the sub-scale showed that the highest level of satisfaction in quality of life occurred among mothers from rural areas, particularly mothers of girls in the physical health domain and mothers of boys in the psychological domain. The quality of life of parents of patients with MMC is significantly worse than healthy people in all aspects (physical health, psychological, environment, and social relationships). Deterioration in the quality of life of mothers with sick children is more common among those living in the city.

Authors' contributions/Wkład autorów

BO-Z – study design, data collection and interpretation, literature search. JW – data collection and interpretation,

acceptance of final manuscript version. WK – data collection and interpretation, statistical analysis, literature search.

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Conflict of interest/Konflikt interesu

None declared.

Ethics/Etyka

The work described in this article have been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; EU Directive 2010/63/EU for animal experiments; Uniform Requirements for manuscripts submitted to Biomedical journals.

The own research were conducted according to the Good Clinical Practice guidelines and accepted by local Bioethics Committee, all patients agreed in writing to participation and these researches.

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